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A Clinical Study on Intrahepatic Biliary Cystadenoma and Cystadenocarcinoma

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Background/Aims: Biliary cystadenomas and cystadenocarcinomas are rare intrahepatic cystic neoplasms that are pathologically characterized by multilocular appearances. We analyzed the clinical pathologic and radiologic features of these rare diseases. **Methods:** Nine patients with intrahepatic biliary cystadenoma (n=6) or cystadenocarcinoma (n=3) which was surgically proven were included in this retrospective study. **Results:** There were 8 women and one man, with the mean age of 41 years (range, 23-60 years). Abdominal pain and/or distension were the major clinical symptoms. The appearance of the tumors was either unilocular cystic or multilocular cystic. The tumors did not communicate with bile ducts. Papillary excrescences, nodular thickening of internal septa and mural nodules were seen in one case of cystadenoma and in all three cases of cystadenocarcinomas. Pathologically, these tumors were typically well-encapsulated and composed of multiple cysts that were lined with biliary type epithelium. After complete resection, all patients except one who had positive lymph nodes did not experience recurrence of tumor. **Conclusions:** When a well-encapsulated, multilocular intrahepatic cystic mass is detected, especially in a middle-aged woman, biliary cystadenoma and cystadenocarcinoma should be suspected and resected. (**Kor J Gastroenterol 1999;33:417 - 424**)

Key Words: Intrahepatic biliary cystadenoma, Cystadenocarcinoma

1.

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5% .1 1892 , 23 60

150 41 6

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211가 Choi 11 51 .

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3 (2, 8, 9),

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6 3 (8)(Table 2).

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2.

AST ALT

Table 1. Age, Sex, Pre-operative and Final Diagnosis

Case	Sex	Age	Pre-operative diagnosis	Final diagnosis
1	F	51	BC	BC
2	F	27	Simple cyst	BC
3	F	30	Cystic mass	BC
4	F	31	BC	BC
5	F	52	BC	BC
6	F	23	Cystic mass	BC
7	F	50	Cholangiocarcinoma	BCA
8	F	60	Cavernous hemangioma R/O Hepatocellular carcinoma	BCA
9	M	43	BC	BCA

BC, biliary cystadenoma; BCA, biliary cystadenocarcinoma; Case 3 and 7 were previously reported by Kim and Park (reference 2).

Table 2. Clinical Findings of the Patients

Case	Chief complaint	Duration	Symptoms	Physical examination
1	Abdominal fullness	8 months	Indigestion, fatigue	Unremarkable
2	Epigastric pain	38 months	Colicky pain	Unremarkable
3	RUQ mass	12 months	Indigestion, weight loss	Palpable mass
4	Diarrhea	3 months	Fatigue	Tenderness on RUQ
5	RUQ mass	18 months	Constipation	Palpable mass
6	Weight loss	12 months	Indigestion	Palpable mass
7	RUQ mass	2 months	Indigestion	Palpable mass
8	Epigastric pain	7 days	Indigestion	Jaundice, tenderness
9	Epigastric pain	3 days	Indigestion	Unremarkable

RUQ, right upper quadrant; Case 2 had symptomatic gallstone disease.

Table 3. Laboratory Findings of the Patients

Case	AST (IU/L)	ALT (IU/L)	ALP (IU/L)	-GT (IU/L)	Total bilirubin (mg/dL)	CEA (ng/ml)	FP (ng/ml)
1	21	25	77	59	1	1.3	1
2	12	21	51	-	0.5	-	3
3	11	7	39	-	0.7	-	1
4	8	10	57	17	0.8	0.1	2
5	23	26	91	43	1.8	2.4	4.6
6	16	25	52	-	1	1	1
7	28	10	90	-	0.8	-	1
8	142	278	250	112	4.7	2.9	3
9	17	11	117	75	0.6	1.1	1

AST, aspartic transaminase; ALT, alanine transaminase; ALP, alkaline phosphatase; -GT, gamma-glutamyl transferase; CEA, carcinoembryonic antigen; FP, alpha-fetoprotein.

1 (8) 3 , 6 . 6
 , alkaline phosphatase 2 , 4 가
 3 (5, 8, 9) , -GTP 4 (1, 5, 8, 9) 3 1 2
 . 1
 1 (8) (7) ,
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 carcinoembryonic antigen (CEA) alpha- (multilocular) (septa) 가
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 (Table 3). 3
 (7, 8, 9).
 3. (ERCP) 4
 (1, 3,

Table 4. Radiologic Findings of the Patients

Case	Radiologic tools	Anatomic site	Number of mass	Well defined	Multilocular or septa	Internal echo	Calcification	IHD dilatation	Communication (ERCP)
1	US, CT ERCP, FNAC	Left	1	+	+	-	-	-	-
2	US	Right	1	+	-	-	-	-	ND
3	US, CT, ERCP, Angio	Right	1	+	+	-	-	-	-
4	US, CT	Left	1	+	-	+	-	-	ND
5	US, CT, ERCP	Left	1	+	+	+	-	-	-
6	US, CT	Left	1	+	+	+	-	-	ND
7	US, CT, Ga scan	Left	Multiple	+	+	+	-	-	ND
8	US, CT, Angio	Right	1	+	+	-	-	-	ND
9	US, CT, ERCP, FNAC	Left	1	+	+	+	-	-	-

US, ultrasonography; CT, computed tomography; ERCP, endoscopic retrograde cholangiopancreatography; FNAC, fine needle aspiration cytology; Angio, angiography; Ga scan, gallium scan; ND, not done.

Table 5. Pathologic Findings of the Patients

Case	Anatomic site	Size (cm)	Cut surface	Lining	Pedicle	Content	CMS
1	Left	10×8×3	Multilocular	Irregular thickening but smooth	-	Mucinous	-
2	Right medial	2.5×2.5×2.5	Unilocular	Smooth	-	Bile	-
3	Right inf-med	10×10×8	Multilocular	Smooth, trabecular, papillary	-	Mucinous	+
4	Left	6×4×4	Unilocular	Granuloma	-	Mucinous	-
5	Left	12×18×8	Unilocular	Nodular	-	Chocolate	+
6	Left medial	6×5	Multilocular	Smooth, Trabecular	-	Mucinous	+
7	Left	6×5×2	Multilocular	Papillary	-	Greyish	-
8	Right post-inf	10×9×4	Unilocular	Papillary	-	Whitish	-
9	Left lateral	6×8×4	Unilocular	Papillary	-	Mucinous	-

CMS, Cystadenoma with mesenchymal stroma.

5,9).

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(3, 8)(Table 4).
4.
9 cm (2.5-18 cm)
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6 4
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(pedicle)
, 1 (2)
,
(cysta-
denoma with mesenchymal stroma, CMS)
3 (3, 5, 6).

(Table 5).
5.
5 , 1
(5),
(7, 8, 9)
1 (7), 1
4 (8),
1 3
(9)(Table 6).

1892 12 150 가
가
1989 2 가

Table 6. Treatment and Prognosis

Case	Treatment	Recurrence
1	Excision	-
2	Excision	-
3	Excision	-
4	Excision	-
5	Lobectomy	-
6	Excision	-
7	Lobectomy	+
8	Lobectomy	-
9	Lobectomy	-

3-11
3 , 13
1
9 9 ,
16 ,
(cysta-
denoma with mesenchymal stroma, CMS)
30-50 가 가
47 가 , 60 가
.1,13 4 50
23 52
35.8 51
10
, 9 8
가 ,
,
.13
2-25 cm

가
가

.1,11,13 가
3 가 가 4 .
가
2.5-18 cm
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2.5
cm
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alkaline phosphatase -GTP 1 (2)
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5
2 4
CEA 2
FP , 2
4
.14,17
가 가 ,
가 .13,15
1
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가 ,7,18,19
85%가
.14,17 , 15%가
50%, 29%,
16% 2

, .423
.113
6 2 , 4 가 .132024
, 3 5 가 1
1 , 2
가 . 3 ,
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가 .

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: 9

CMS,
COMS (cystadenoma without mesenchymal
stroma) .2022 : 20

423

CMS 6 3 ,
, , 가
17 가 CMS가 . : 41

.202123

15 가 ,

6 3
3 . 9 cm

가 ,

.23 가 가
, , (marsu-
pialization), , (sclerosis)

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